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### CASE REPORT

## Intraoral venous malformation with phleboliths

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#### KEYWORDS

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**Abstract** The most common type of vascular malformation is the venous malformation and these are occasionally associated with phleboliths. We report a case of a 45 year old woman with intraoral venous malformation with phleboliths.

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#### 1. Introduction

A hemangioma is a tumor of infancy that undergoes a phase of rapid growth and expansion followed by a period of slow but steady regression during childhood. In contrast, vascular malformations are congenital collections of abnormally formed channels that are present at birth, grow proportionally with the child and do not undergo regression (Beck and Gosain, 2009). They can be divided into two groups depending on their hemodynamic and lymphodynamic characteristics: low flow and high flow malformations. Low flow lesions include capillary, venous and lymphatic malformations or a combination of these elements. High flow lesions include arterial malforma-

tions and arterio-venous malformations (Burns et al., 2009). Approximately 40% of the venous malformations are located in the head and neck region and these occasionally are associated with phleboliths. Therefore, these calcifications are considered typical features of venous malformations. In addition, they are usually visible on plain radiographs (Scolozzi et al., 2003). We report a patient who presented with an intraoral venous malformation associated with phleboliths.

#### 2. Case report

A 45 year old woman reported to the department of Oral Medicine and Radiology, Kothiwal Dental College and Research Centre, Moradabad, India, with a chief complaint of gingival bleeding on brushing. An intraoral examination revealed calculus deposition along with gingival inflammation, for which the patient was scheduled for oral prophylaxis. A painless bluish purple discoloration, irregular in shape and measuring approximately 3 × 2 cm, was seen on the labial mucosa and vestibule, extending from the region of right maxillary central incisor to first premolar Fig. 1. It was present since birth and increased progressively with age. Blanching on applied pressure was noted. There were no visible pulsations neither was there any bruit felt. These clinical findings were strongly suggestive of a vascular malformation. A maxillary anterior topographic occlusal radiograph showed two round to oval radiopaque bodies located labial to the premolars Fig. 2. These

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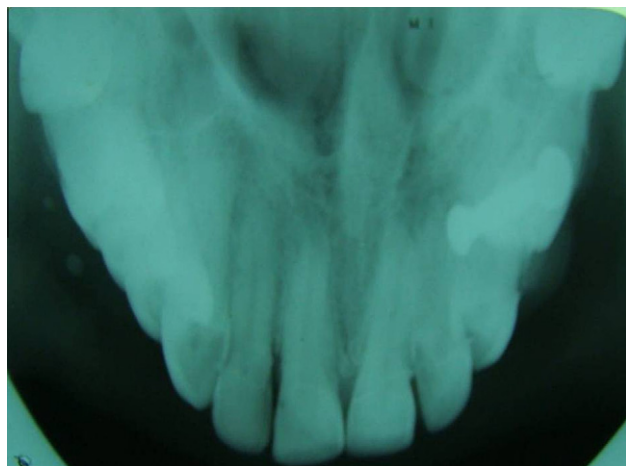
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**Figure 1** An intraoral photograph showing the venous malformation.

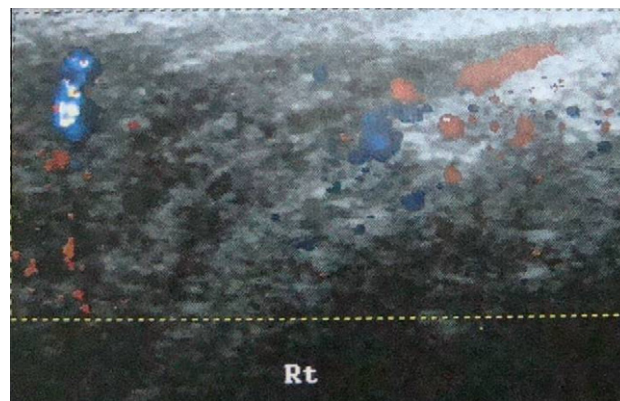


**Figure 2** A maxillary anterior topographic occlusal radiograph showing two round radiopacities in the buccal mucosa.

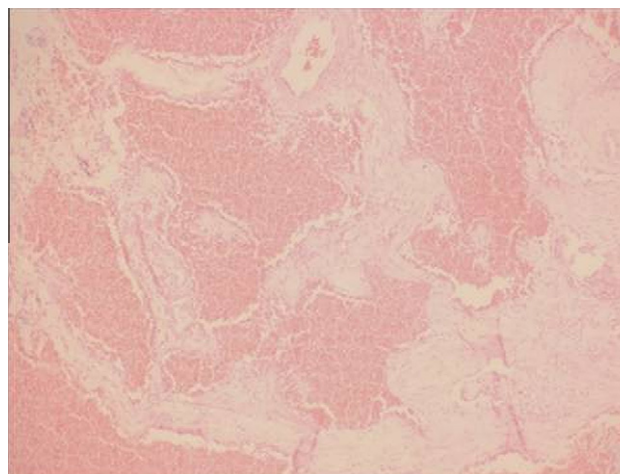
were interpreted as phleboliths. Color Doppler ultrasonography revealed a solid hypoechoic lesion on the upper labial mucosa with well defined margins measuring  $27.9 \times 6.2 \times 2.1$  mm. Few foci of calcifications were seen within the lesion, with the largest measuring 2.2 mm. Peripheral vascularity was seen in the lesion with both arterial and venous patterns [Fig. 3](#). A biopsy was performed under local anesthesia. Two round to oval calcifications were obtained together with a portion of the mucosal lesion. A histopathological examination disclosed several thin walled irregular venous channels lined by flattened endothelium supported by dense uninflamed fibrous connective tissue stroma [Fig. 4](#). Microscopically, the calcifications were consistent with phleboliths. The case was diagnosed as intraoral venous malformation with phleboliths.

### 3. Discussion

Venous malformations are slow growing vascular malformations that are present at birth. They are non proliferating vascular birthmarks composed of anomalous ectatic venous channels. A variety of terms including “venous angioma”,



**Figure 3** Color Doppler showing a solid hypoechoic lesion on the upper labial mucosa with two foci of calcification within the lesion. Peripheral vascularity within the lesion with both arterial and venous patterns can also be appreciated.



**Figure 4** Histopathological examination using Hematoxylin and Eosin stain with 40x magnification, revealed thin walled irregular venous channels lined by flattened endothelium.

“cavernous angioma”, “cavernous hemangioma” and “phleboangioma” have been used in literature to describe these anomalies ([Garzon et al., 2007](#)).

Vascular malformations are present at birth but are not always evident. They typically become more prominent as the patient matures; the most pronounced enlargement is usually seen from infancy to puberty with less pronounced changes in adulthood. They usually present as soft, compressible blue masses. The blue color is pathognomonic and is caused by the presence of ectatic anomalous venous channels within the dermis. There is no increase in the local skin temperature or thrill when the lesion is palpated ([Garzon et al., 2007](#)). Their typical slow growth may cause them to be asymptomatic for many years. Infection or trauma may cause these lesions to acutely increase in size ([Rosbe et al., 2010](#)). They may also sometimes expand in response to hormonal changes (e.g., puberty and pregnancy) ([Tan et al., 2004](#)).

The head and neck region is most frequently involved (60%), followed by 25% located on the trunk and 15% on the extremities ([Finn et al., 1983](#)). Venous malformations can occasionally be completely intraosseous and the mandible is

the most common bone involved, although maxillary, nasal, and frontal lesions have also been reported (Ethunanadan and Mellor, 2006). Facial vascular malformations involve the skin and subcutaneous layers, but often have extension into muscle and oral mucosa (Garzon et al., 2007).

Venous malformations typically expand after the Valsalva manoeuvre and may be flattened with applied pressure (Dubois et al., 2001).

When there is no frank bluish hue and the overlying skin appears normal, a venous malformation must be differentiated from the high flow vascular malformations and hemangiomas: other possible differential diagnosis includes lymphangiomas, resolving hematomas, or neoplastic conditions (Trop et al., 1999).

Complications of the vascular malformations in the head and neck area and mucosal surface include significant cosmetic defects, recurrent bleeding, obstruction of airway and interference with normal speech and dentition (Beck and Gosain, 2009).

Doppler ultrasound (US) is essential in differentiating venous malformations from other vascular anomalies. US should be performed with a high-frequency linear array transducer (5–10 MHz). Exploration begins with a gray-scale examination to delineate the margins of the malformation. Venous malformations appear as hypochoic or heterogeneous lesions in 80% of cases. Anechoic channels can be visualized in less than 50% of cases (Dubois et al., 2001).

As with all vascular malformations, conventional hematoxylin-eosin staining techniques for venous malformations (VMs) reveal irregular variably dilated or thickened dysplastic-appearing vascular channels lined with flat mature endothelial cells in contrast to hypercellularity seen in vascular tumors (Legiehn and Heran, 2008). These vascular spaces are usually filled with an abundance of erythrocytes. Capillaries and venules may reside within the VM substance. In addition to the absence of internal elastic lamina, there is a relative paucity or intermittent absence of smooth muscle within the VM channel wall with occasional locules of disorganized smooth muscle identified emanating from the vascular wall into the surrounding stroma. Localized intravascular coagulopathy is frequently present within VMs and as a result, luminal thrombi can develop and become calcified and form phleboliths (Legiehn and Heran, 2008).

Elastic stains (elastic Van Gieson stain) are helpful in distinguishing between arteries and veins. Negative GLUT-1 staining (erythrocyte type glucose transporter protein-1) may facilitate distinction from juvenile hemangioma (Leon-Villapalos et al., 2005).

Phleboliths associated with vascular anomalies were initially found in the splenic vein by Canstatt in 1843 and in the maxillofacial region by Kirmission in 1905. According to Ribbert's theory phlebolith formation begins with intravascular thrombus formation and is followed by progressive lamellar fibrosis. Calcium phosphate and calcium carbonate are deposited at the center of the thrombus, with an extension of mineralization to the periphery. Microscopically phleboliths consist of calculi with characteristic concentric lamination. Radiographically, they present as multiple round and oval laminated bodies with either radiolucent or radiopaque cores (Scolozzi et al., 2003).

Though cases of intraoral phleboliths have been reported in the literature previously, but, their association so far, has been noted predominantly with hemangiomas (Ikegami and Nishijima, 1984; Sano et al., 1988; Atluğ et al., 2007). A

medline search using the words 'venous malformation' and 'intraoral phleboliths' revealed only one case report by Scolozzi et al. (2003). They reported intraoral venous malformation in the left posterior buccal mucosa and retromolar trigone in a 92 year old woman with multiple round to oval phleboliths.

Salivary calculi should be considered in the differential diagnosis. They are typically uniformly radiopaque and tend to have a ductal anatomic location and elongated shape. Sialography reveals a filling defect at the site of salivary calculus, whereas phleboliths appear to be external to the duct system (Scolozzi et al., 2003).

Superficial venous malformations typically present as a soft, compressible, bluish mass. However, these features may not be fully apparent in intramuscular or deeply situated lesions. It is important to be aware that the radiographic finding of phleboliths in the soft tissues of head and neck region constitutes evidence of the presence of a vascular malformation (Scolozzi et al., 2003).

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